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CASE REPORT

Dupuytren contracture after acute traumatic hand injury in an adolescent: A case report

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ABSTRACT

Dupuytren's disease is a fibroproliferative disease that affects the palmar hand, causing progressive, permanent, and symptomatic flexion contracture of the digits. It is a disorder of polygenetic and multifactorial origin, usually affecting middle-aged males. Dupuytren's disease developing as a result of hand trauma in younger ages is rarely reported. In this case, we present a 14-year-old male patient with Dupuytren's disease following acute traumatic injury. We performed the fasciectomy and Z-plasty with full thickness skin graft. He recovered completely additional injection of triamcinolone for one year.

Keywords: Adolescent, dupuytren contracture, hand injuries, triamcinolone.

Dupuytren's disease is a chronic progressive fibroproliferative hand disorder of the palmar fascia, which may lead to an irreducible and disabling progressive flexion of the fingers.[1] The standard course of this disease manifests as hard nodules in the palmar aponeurosis, leading to the formation of longitudinal fibrous bands.[2] The cause of Dupuytren's disease is complex and multifactorial. Genetic association with the disease is well known, and recent studies proposed association between Dupuytren's disease and nongenetic factors including diabetes mellitus, alcohol, smoking, liver disease, and repetitive use of the hand.[3-5] A specific form of Dupuytren's contracture developing as a result of hand trauma has been discussed in many studies.[6-8] There is still no definite conclusion about the association between traumatic events and the appearance of Dupuytren's contracture. However, Dupuytren's disease following acute injury that eventually leads to surgical intervention in younger patients is relatively rare. Herein, we report a rare case of Dupuytren's disease in a younger patient following traumatic injury.

CASE REPORT

A 14-year old-male patient was referred to plastic surgery with hand injury by sharp metallic fence while slipping down. On physical examination, a wound about 3 cm in length was found at the right fourth metacarpophalangeal (MCP) joint area. The wound was superficial to the subcutaneous layer, and tendon injury was absent. The patient had no difficulty in fully extending and flexing his fingers, and showed no neurologic symptom. The laceration wound was repaired using nylon 5-0 suture.

The patient was lost to follow-up, and one year after the trauma, the patient revisited our department

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with a limitation of motion in the MCP joint crease area. The patient had an extension deformity of 40° at fourth MCP joint level. Physical examination revealed about a prominent 6.0×0.5 cm palmar fibrous band over the metacarpal area extending to the middle interphalangeal joint (Figure 1).

The tabletop test was positive, which revealed the possibility of Dupuytren's contracture. The patient had no predisposing risk factor for the disease and was an ordinary student who did not perform any intense hand work. The patient with a positive history of acute trauma was evaluated by the Elliot and Regoowansi's[8] criteria for recognition of Dupuytren's contracture after acute injury.

Intralesional injections of triamcinolone acetonide into the palmar nodule was done for a total of four times during three months but showed no improvement in the contractures. Under general anesthesia, we performed fasciectomy on the approximately 6.0×0.5 cm-sized cord-like scar band, and a 5 cm pretendinous cord was revealed above the flexor tendon sheath. Under loupe magnification, *en bloc* resection of the cord was done (Figures 2, 3). After *en bloc* resection of the scar band and pretendinous cord, full motion of fourth finger was confirmed. A full thickness skin graft was done for the coverage of the 6.0×0.5 cm-sized palmar defect. After coverage, additional multiple Z-plasty was performed from the palmar area to the middle phalangeal area to minimize tension (Figure 4). The skin incision was closed with a simple interrupted suture with 5-0 nylon sutures.

In the histological examination, multiple collagen fibers and myofibroblasts by the collagen degeneration were shown as the scar contractures (Figure 5).

The patient was discharged after one week without any complications. From two months

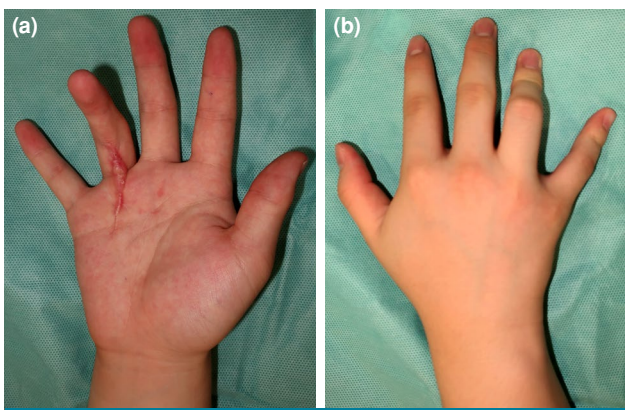

FIGURE 1. (a, b) Initial clinical photograph of band formation in the proximal phalangeal area of the right ring finger.

FIGURE 2. Intraoperative clinical photograph of the pretendinous band involving the skin.

FIGURE 3. Perioperative photograph of the excised pretendinous band and skin.

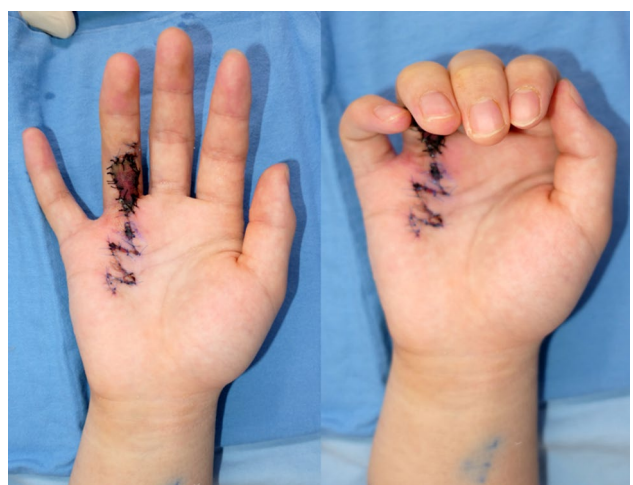

FIGURE 4. Postoperative photograph of the full thickness skin graft and multiple Z-plasty.

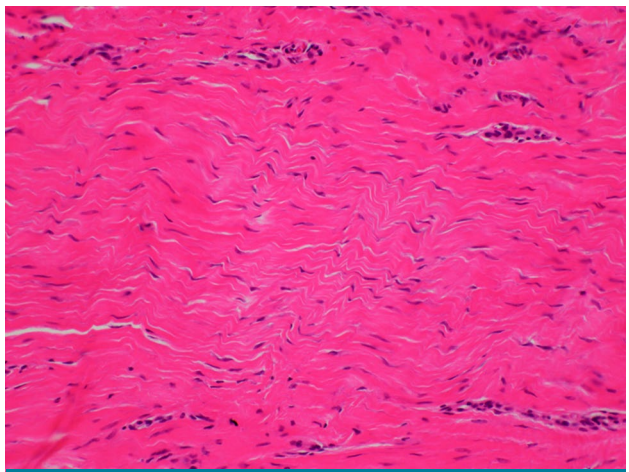

FIGURE 5. Multiple collagen fibers and myofibroblasts were shown by the collagen degeneration (H&E, ×100).

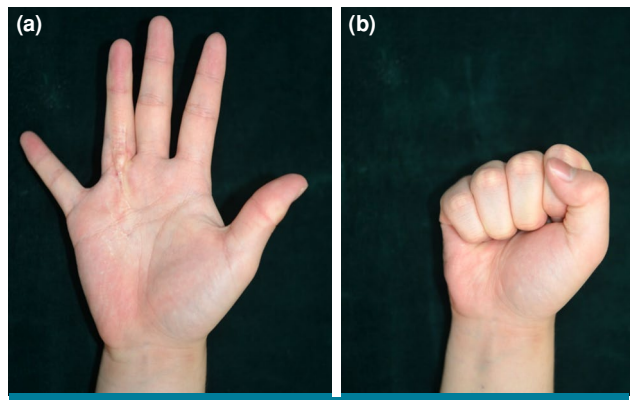

FIGURE 6. (a, b) The patient's right hand six months after surgery without any complications.

after the surgery, a total of six times of additional triamcinolone injection was done for one year. The two-year postoperative follow-up demonstrated full extension of all digits in the right hand and showed no sign of recurrence (Figure 6).

DISCUSSION

Dupuytren's disesase is a fibrosing disorder of the palmar fascia characterized by the formation of a nodule, which involves into a cord. It is known as a hereditary disease preferentially affecting Caucasians originating from Northern Europe.[9] However, its genetic mode of inheritance is not well understood. Beyond genetic cause, some environmental factors, including alcohol consumption, tobacco exposure, diabetes, and epilepsy, are known to be associated with the disease.[1,4,5] It commonly affects males between the ages of 40 and 80, very rarely found in younger ages.[1-4]

Until recently, many studies have been reported to determine if Dupuytren's disease following acute injury runs a different course from typical Dupuytren's disease. In 1891, Anderson[10] first introduced the term "false Dupuytren's contracture-traumatic from," which he claimed is an entirely different and milder condition from true Dupuytren's disease. In 2005, Elliot and Ragoowansi[8] suggested the criteria for recognition of Dupuytren's contracture after acute injury. More recently, Rayan and Moore[11] proposed a new entity "non-Dupuytren's palmar fascial disease." However, some studies suggest that although Dupuytren's disease after injury follows a different course, which is less progressive and less extensive, rarely requiring surgery, and with a radial shift of presentation of the disease, it should be considered as a subtype of Dupuytren's disease, not a separate one.[5-7]

Pathogenesis of Dupuytren's disease is still unfolding. A major hypothesis suggests that the initiation of the disease includes altered immune responses, local hypoxia, and abnormal wound healing responses.[4] Brauns et al.[12] claimed that repetitive trauma leads to nerve growth factors to be released, which promote the migration and proliferation of fibroblast-like cells, then occurs thickened nodules and cords, which present as Dupuytren's contracture.

This case is very unique in that Dupuytren's disease after acute trauma injury is rarely reported in teenagers. Furthermore, unlike most cases of Dupuytren's contracture after trauma that do not require surgical intervention, our patient showed no progress with triamcinolone injection, which eventually led to the surgery. Clinicians should be aware that only a single minor traumatic event can lead to Dupuytren's disease, even in younger ages.[10]

In conclusion this case showed that a single traumatic event can lead to Dupuytren's disease even if in younger ages without any predisposing factors. Further elucidation of the mechanisms involved in Duptuytren's disease after traumatic injury is required for an early diagnosis and adequate treatment.

Patient Consent for Publication: A written informed consent was obtained from the parent of the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept, design, writing the article, references and fundings: J.Y.C.; Control/supervision: B.F.S.; Data collection and/or processing, analysis and/or interpretation: Y.J.C.; Literature review: S.N.J.; Critical review: B.F.S.; Materials: Y.J.C.

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